

Accepted Manuscript

Title: Diagnostic accuracy of the spanish version of the paediatric sleep questionnaire for screening of obstructive sleep apnea in habitually snoring children.

Author: Katalina Bertran, Tomas Mesa, Karina Rosso, Maria José Krakowiak, Eduardo Pincheira, Pablo E. Brockmann

PII: S1389-9457(15)00084-2
DOI: <http://dx.doi.org/doi: 10.1016/j.sleep.2014.10.024>
Reference: SLEEP 2668

To appear in: *Sleep Medicine*

Received date: 20-8-2014
Revised date: 16-10-2014
Accepted date: 17-10-2014

Please cite this article as: Katalina Bertran, Tomas Mesa, Karina Rosso, Maria José Krakowiak, Eduardo Pincheira, Pablo E. Brockmann, Diagnostic accuracy of the spanish version of the paediatric sleep questionnaire for screening of obstructive sleep apnea in habitually snoring children., *Sleep Medicine* (2015), <http://dx.doi.org/doi: 10.1016/j.sleep.2014.10.024>.

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.



**Diagnostic accuracy of the Spanish version of the Paediatric Sleep Questionnaire
for screening of obstructive sleep apnea in habitually snoring children.**

Katalina Bertran¹, Tomas Mesa^{1,2}, Karina Rosso²,
Maria José Krakowiak², Eduardo Pincheira², Pablo E. Brockmann^{1,2}

Affiliations:

- (1) Department of Pediatric Cardiology and Pulmonology, Division of Pediatrics,
School of Medicine, Pontificia Universidad Católica de Chile, Santiago, Chile.
- (2) Sleep Medicine Center, Department of Neurology, School of Medicine,
Pontificia Universidad Católica de Chile, Santiago, Chile.

Corresponding author

Pablo E. Brockmann, MD, PhD

Pediatric Pulmonologist, Sleep Medicine, Professor Assistant of the Department of
Pediatrics, School of Medicine, Pontificia Universidad Católica de Chile

Lira 85 5to piso, 330074 Santiago de Chile, Tel: 56.2.23543767

pbrockmann@med.puc.cl

Keywords: Snoring, apnea, diagnostic, accuracy, questionnaire, polysomnography.

Highlights:

- The Spanish version of the PSQ showed a good diagnostic test accuracy for screening OSA in children.
- Six items concerning respiratory symptoms showed the highest diagnostic accuracy.
- This subscale score identified correctly 89% of the children with OSA.

Accepted Manuscript

ABSTRACT

Objectives: We aimed to determine the diagnostic test accuracy of the Spanish version of the respiratory symptoms scale of the Pediatric Sleep Questionnaire (PSQ) in habitually snoring children for identifying obstructive sleep apnea (OSA).

Methods: Habitually snoring children referred for polysomnography (PSG) were recruited. Parents answered the PSQ prior to PSG. According to an apnea hypopnea index >1.0 in PSG children were divided into OSA and primary snorers. Correlations to PSG indices and diagnostic test accuracy measures were calculated.

Results: Of the $n=83$ ($n=53$ males, mean age 9.5 ± 3.6 years) habitually snoring children included, $n=35$ had OSA. The previously validated PSQ cut-off value of 0.33 showed a specificity of 0.72 and sensibility of 0.78. The PSQ score correlated significantly with the apnea hypopnea index $r_s = 0.313$ ($p\text{-value} = 0.004$). Six items of the PSQ were significantly different between cases and controls. A subscale constructed on these 6 PSQ items concerning respiratory symptoms showed a good sensitivity (0.886), and an excellent negative likelihood ratio (0.261). PSQ was able to identify correctly 89% of the children with OSA.

Conclusions: This version of the PSQ was able to identify children with OSA, separating them from those with primary snoring. Use of this simple, standardized questionnaire tool seems to be helpful and may improve clinical decision-making in habitually snoring children.

INTRODUCTION

Sleep disordered breathing (SDB) in children is characterized by habitual snoring and a number of daytime symptoms like hyperactivity, somnolence and poor school performance [1]. Obstructive sleep apnea (OSA) is considered the most severe form of SDB, and is defined as a disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction (obstructive apnea) that disrupts normal ventilation during sleep and normal sleep patterns accompanied by symptoms as excessive daytime sleepiness, behavioral problems, learning disabilities, growth retardation or failure to thrive [2] [3].

However, most children who have OSA remain undiagnosed [4]. In pediatric population, snoring prevalence is 3-12 % and OSA is 0,7-2%, mainly associated to adenotonsillar hypertrophy [5, 6].

In general, on OSA evaluation, a good history and physical examination are required. However, OSA may be not diagnosed only based on clinical history or physical examination [7]. The gold standard for the diagnosis of OSA is sleep laboratory-based polysomnography (PSG)[8]. Despite its diagnostic advantages, PSG is an expensive, time consuming assessment. Considering the high prevalence of OSA and the need for prompt treatment, simpler screening tests seem to be urgently necessary. However, there is currently no simple, cost-efficient and accurate screening test for OSA that helps to identify children, who are at risk for serious OSA-related complications and who would benefit from early detection, PSG and treatment. As a screening option, several questionnaires have emerged as a possible screening tool for OSA and were discussed in detail by Spruyt et al. [9, 10].

Recently, our group published a systematic review on the diagnosis of OSA in children [11]. There were only few tests that achieved good or excellent levels of diagnostic accuracy [11]. Among these tests, a simple questionnaire achieved a reasonable diagnostic accuracy: the Paediatric Sleep Questionnaire (PSQ) [3].

Chervin et al. developed and reported the validity and reliability of the PSQ including three symptoms: snoring, sleepiness and behavioural problems [3]. The usefulness of the PSQ was proven in children in whom OSA was suspected and compared with non-snoring controls [3]. That study concluded that the PSQ had an excellent

sensitivity and specificity for identifying children with OSA, when applied to a group of referred snoring children and also to a group of controls [3].

Vila et al. translated the PSQ into Spanish and determined its reliability by test-retest and internal consistency methods among a sample of 99 patients [12]. Ninety-one percent of the questions showed good concordance, following the criteria established in the test-retest method, concluding that the Spanish version of the PSQ could be a useful tool for epidemiological research.

Therefore, we aimed to perform a diagnostic test accuracy study for this version of the PSQ, comparing it with full sleep-lab based PSG in children. Furthermore, we aimed to analyze the screening usefulness of this version of the PSQ for diagnosing OSA with currently accepted definitions exclusively in habitually snoring children referred to PSG.

Accepted Manuscript

METHODS

Subjects

We included all children aged 0 – 15 years who were referred to PSG between 2007 to 2012 to the Sleep Laboratory of the Pontificia Universidad Católica de Chile, Santiago, Chile. Only habitually snoring children referred to PSG for diagnosing OSA were selected for this study. These habitually snoring children were sent based on the clinical judgment of their physicians to perform a PSG, no prior screening tool was used for their selection.

PSG was performed in patients, with no acute respiratory infections. Nineteen patients had history of asthma symptoms and one had allergic rhinitis. Of them, n=7 patients were on inhaled corticosteroids, n=4 on antiasthmatics and n=14 on antihistaminics.

PSGs and their corresponding PSQs were revised. Nutritional status was assessed using body mass index (BMI, kg/m²). Obesity was diagnosed if the BMI was >2 z-scores over the mean reference value for age and gender [13]. We excluded children with any chronic neurological or genetic diseases, cerebral palsy, and Down syndrome. This study was approved by the ethical board of Pontificia Universidad Católica de Chile (IRB approval number 12-317).

Pediatric Sleep Questionnaire

The evening of PSG, parents completed the Spanish version of the PSQ [12]. The sleep related breathing disorders scale of the PSQ [14] was analysed for the purposes of the current study. This scale contains 22 questions about symptoms including snoring characteristics, as duration, intensity, frequency, episodes of apnea, mouth breathing, presence of enuresis, excessive sleepiness during the day, headache, weight

and height percentile, symptoms of hyperactivity-impulsivity and inattention. Possible responses for each item were “yes” = 1, “no” = 0, “don't know” = missing. To reduce responses to yes/no formats in those items with more possible answers, “does not apply” and “applies just a little” were also scored as no (0), whereas “applies quite a bit” and “definitely applies most of the time” were scored as yes (1)[3]. The PSQ score was calculated by summing total count of all “yes” answers / total count of all “no” and “yes” answers. We used the previously validated cut-off value of 0.33, which would be most effective in identifying paediatric OSA [3, 14]. In addition to that, we aimed to analyse a subscale based on those items that showed the highest representation within our sample. The PSQ score was calculated blindly to PSG analysis.

Polysomnography

PSG was conducted in the sleep lab during night using a computerised polysomnographic system (ALICE 5.0, Respironics, Andover, MA, USA) according to current international standards published by the American Academy of Sleep Medicine [15]. The PSG was performed in the sleep lab during night with continuous attendance. The study montage comprised the following channels: 3-lead electroencephalography, 2-lead electrooculography, 3-lead submental is electromyography, chest and abdominal wall movements, nasal pressure transducer, snoring, pulse oximetry-derived arterial hemoglobin oxygen saturation and pulse waveform, electrocardiogram and instantaneous beat-to-beat heart rate, digital audio and video. Respiratory events and sleep architecture were analyzed according to the above-mentioned criteria [15]. Arousals were identified as defined by the American Sleep Disorders Association Task Force report [16]. Central, obstructive, and mixed

apneas and hypopneas were identified according to current recommendations [17]. Obstructive apneas were defined as the absence of airflow with continued chest wall and abdominal movement for duration of at least two breaths. Central apneas were defined as the absence of nasal flow and thoraco-abdominal movements for >20 seconds, or for > 2 breaths if the episode is accompanied by desaturation or arousal. Hypopneas are defined as a decrease in nasal flow of at least 30% with a corresponding decrease in SpO₂ of 3% or more and/or an arousal [17]. The apnea–hypopnea index (AHI) was defined as the number of obstructive and mixed apneas and hypopneas per hour of total sleep time. OSA was defined as an AHI ≥ 1 . Based on this definition the subjects were classified as having OSA (i.e., AHI ≥ 1) or being a primary snorer (AHI < 1). Analysis of PSG was conducted blinded to PSQ results.

Statistics

Descriptive statistics (mean, standard deviation [SD] for normally, and median, minimum, maximum for non-normally distributed variables) were used to outline subject characteristics, PSQ and PSG. Comparisons between OSA and primary snorers were made with the non-parametric Mann-Whitney-U test. Correlations between diagnostic test results and PSG indices were calculated using Spearman's rank correlation coefficient r_s . Correlations > 0.3 were defined as “acceptable”. Subjects with missing PSQ or PSG data were excluded from the study.

A p-value of < 0.05 was considered statistically significant.

In order to select those items chosen for the subscale, we performed a factor analysis, by the main components method with varimax rotation, and analysis of variance. Those items with the highest load and that performed significantly different in children with OSA versus primary snorers were selected.

In order to calculate the diagnostic test accuracy of continuous diagnostic test parameters, receiver-operating characteristic (ROC) curves were developed and the area under the ROC curve (AUC) as well as its 95% confidence intervals (95%CI) calculated. Based on cut-off values and 2 x 2 tables, sensitivity, specificity, as well as positive and negative likelihood ratios were calculated. Sensitivity and specificity >80% were defined as “acceptable” [11, 18]. Index tests with a positive likelihood

ratio > 3 *and* a negative likelihood ratio < 0.3 were defined as having “good” diagnostic test accuracy (DTA) [11].

Accepted Manuscript

Results

Of a total of $n=86$ habitually snoring children, $n=83$ ($n=53$ males) had complete PSG and PSQ data and were included for analysis. Mean (\pm SD) age was 9.5 ± 3.6 years. There were 15 (20.5 %) obese children in the total sample. According to the AHI, in $n=35$ children an OSA was diagnosed. Table 1 gives demographic and PSG details for the total sample, children with OSA, and primary snorers. Of the 22 items of the PSQ scale following 6 questions on respiratory symptoms were significantly different between cases and controls: “always snores”, “snores loudly”, “trouble breathing”, “observed apneas”, “mouth open during day”, and “unrefreshed in morning” (Table 2). These 6 significant items were used as a subscale for further calculation.

Correlations between PSQ questionnaire score and PSG indices

With the previously established cut-off value of 0.33, applied to the entire sample, the PSQ score showed a significant correlation with the AHI: $r_s = 0.313$ (p -value = 0.004). The same figure for the correlations with the arousal index, and mean SpO₂ were 0.107 (p -value = 0.337), and -0.118 (p -value = 0.073), respectively. Table 3 gives the correlations between the PSQ score and all analyzed PSG indices.

Diagnostic test accuracy of the PSQ score

Using 0.33 as cut-off for the PSQ scale, a ROC curve was constructed (Figure 1), obtaining an AUC of 0.687 (95% CI 0.567-0.808). The calculated diagnostic test accuracy measures for this cut-off were: sensitivity 0.714, specificity 0.521, positive predictive value 0.521, negative predictive value 0.714, positive likelihood ratio 1.491, and negative likelihood ratio 0.549. Based on the PSQ score >0.33 , 71% of the children with OSA were correctly identified.

The AUC (95%CI) for the subscale of the 6 significant items was 0.751 (0.644 – 0.858). The cut-off >0.1 showed the best diagnostic test accuracy measures for this subscale: sensitivity 0.886, specificity 0.538, positive predictive value 0.534, negative predictive value 0.840, positive likelihood ratio 1.575 and negative likelihood ratio 0.261. Table 4 shows DTA analysis dichotomised by age group. Highest DTA results were obtained in pre-schoolers (i.e., aged 2-6 years) reaching excellent positive and negative likelihood ratios: 2.250, and <0.0001 , respectively.

Based on this subscale score it was possible to correctly identify 89% of the children with OSA. Figure 1 shows the AUC for both PSQ cut-offs.

Accepted Manuscript

DISCUSSION

The Spanish version of the PSQ showed significant correlations with polysomnographic indices in a sample of habitually snoring children. Diagnostic test accuracy using the standard cut-off was moderate, but using a recalculated subscale score based on respiratory symptoms it improved meaningfully enough for clinical purposes. Items related to respiratory symptoms showed significant correlations with PSG. The use of these items concerning respiratory symptoms of the PSQ exclusively in habitually snoring children was able to separate primary snoring from OSA with acceptable diagnostic test accuracy.

The gold standard for diagnosing OSA in children is PSG [19]. However, the costs of PSG, waiting lists and the high prevalence of habitual snoring [20-22] have led to a search for alternative screening tests for OSA in children [11]. One of the probably most investigated screening tools are questionnaires [3, 11, 14, 23-25]. Likely reasons for the widespread use of questionnaires are the low costs, easy performance and rapid assessment of their scores [26].

One of these questionnaire (i.e., the Pediatric Sleep Questionnaire (PSQ) –with its SRBD (Sleep-related Breathing Disorder scale) has been extensively studied and showed valid psychometric properties [3]. The PSQ cut-off value was set at 0.33 in the original publications [3, 14]. In those studies (a comparison was made between children referred to PSG and confirmed to have OSA versus non-referred children, whose parents were surveyed in general pediatric waiting rooms) this score and the cut-off value of 0.33 showed a sensitivity of 0.81 and a specificity of 0.87 [3]. Instrument performance did not vary with participant age (2 - 18 years). The score showed good internal consistency and test-retest reliability. Since then, the PSQ has been used frequently to assess for OSA risk in research studies and also for clinical purposes. The advantage of the PSQ is surely the easy to calculate score and a precise cut-off that may be used for screening children for OSA. Furthermore, in 2007, Chervin et al. performed a further study that showed that a high PSQ score predicted OSA on PSG with an odds ratio of 2.80 (95%CI: 1.68 - 4.68)[14]. In that study, the previously established cut-off value of ≥ 0.33 showed 0.78 sensitivity and 0.72 specificity. Compared to that results, the Spanish version [12] of the PSQ used in the present study showed similar diagnostic test accuracy. This is interesting, as it demonstrates that even using a different language, the PSQ still is a powerful tool for identifying children at risk for OSA.

In the original validity study however, the non-referred (control) children were not tested to confirm the assumption that they had no OSA [3]. In the present study, we aimed to analyse the performance of the PSQ only in habitually snoring children who were referred to PSG. The fact that all included subjects had the currently accepted gold standard for diagnosing OSA (i.e., PSG) is surely helpful for the purposes of a diagnostic study. Interestingly, the similar diagnostic test accuracy obtained in the present may reflect that PSQ may be an option for separating primary snoring from OSA exclusively in habitually snoring children. In contrast to Chervin's study we did not intend to compare the diagnostic test accuracy in children with habitual snoring versus non-snoring controls. Considering that primary snorers have also severe neurocognitive and cardiovascular consequences [21, 27, 28], screening tools based on the identification of this group should be encouraged in the future. We stress here, that primary snorers should not be considered to be benign [27] and neglected from future screening tools. The importance of these apparently milder forms of SDB, is also emphasised by the strong correlation found between the PSQ score and hypopnea index. This fact may reflect the real importance of hypopneas in children, which seem to be more frequent than apneas [29].

The use of a subscale on respiratory symptoms improved diagnostic test accuracy and sensitivity and specificity. Using a cut-off >0.1 , the negative likelihood ratio of this 6-item subscale was in an "excellent" range. This means that in absence of a positive score in the subscale, the presence of OSA decreases. This cut-off is different from the 0.33 used for the full PSQ scale; however it reflects that even a smaller part of the complete questionnaire could be dichotomised for detecting OSA versus primary snorers. The 6 PSQ items that were selected for inclusion in the present subscale snoring (i.e., "always snores", "snores loudly", "trouble breathing", "observed apneas", "mouth open during day", and "unrefreshed in morning") reflect more respiratory and not behavioural problems associated with OSA. This is interesting, as the original PSQ scale also addressed some issues for hyperactivity and behaviour [3]. However, there is evidence that children with primary snoring have similar behavioural consequences and hyperactivity compared with those with OSA [27, 28, 30]. Then it does not seem surprising that items investigating these consequences may not necessarily be helpful for screening OSA in a sample of habitually snoring children. itself, and possibly some feature of sleep disruption that is not registered in PSG, may lead to the development of associated cognitive and behavioural consequences [30]. This is in line with *Chervin et al* studies on the usefulness of PSQ for prediction of consequences [14]. In that study it was demonstrated that the PSQ score correlated with an attention deficit scale and sleep latency in the multiple sleep latency test [14]. Furthermore, the scale showed even better correlations with those outcomes compared with PSG. In the present study, only the item "unrefreshed in morning" merits special attention, as it was the only

“behavioural” item that was significantly different between cases and controls. One may hypothesise that this item reflecting somnolence might be a specific marker of children with OSA [31-33], but surely this has to be confirmed in future studies intended for that.

Results from the present study are limited due to the retrospective design of the analysis and the lack of OSA- related consequences measurements. This version of the PSQ is not intended to substitute PSG for diagnosing OSA in children. However, the novelty of the use of a simple scale for screening those habitually snoring children with a higher risk for developing OSA seems promising. These results should stimulate future studies in investigating how screening tools (like this questionnaire) may predict OSA-related consequences like hyperactivity, attention deficit, or cardiovascular problems. On the other hand, the current Spanish version of the PSQ was translated using appropriate tools by Villa et al. [12]. In that study, the PSQ has been translated into Spanish with adequate results with respect to validity and reliability [12]. We acknowledge however, that we were far from having used all available psychometric tools to validate this translation and encourage future studies to do so using available guidelines. In one of them, Spruyt et al. have thoroughly investigated sleep questionnaires and have proposed several tools for developing and validating these tools [9, 10]. The evaluation of questionnaires like the PSQ we used requires knowledge of its psychometric characteristics. We intended to validate the PSQ against the currently accepted gold standard (i.e., PSG), but are certain that the psychometric characteristics of this version may still be unknown. However we agree with Spruyt et al. concerning that these questionnaire tools “do not need to be perfect, or even psychometrically exceptional; however, they need to improve clinical decision-making and significantly reduce errors of judgment” [10].

Also, the effect of language and cultural background [10] may have influenced some responses and may explain in some way the differences with the original PSQ [3]. We think this work adds on this point and future studies should continue searching new tools and validate them for various languages, cultures and clinical settings.

Conclusions

The Spanish version of the PSQ was able to differentiate children with OSA from those with primary snoring. Use of this simple, standardize questionnaire tool seems to be helpful for screening and may improve clinical decision-making in habituallysnoring children.

ACKNOWLEDGMENTS

We would like to thank all the children and their parents who-participated in the study, Dr Luis Villarroel for his help with statistics. Also, we thank FONDECYT project number 11130573 for supporting the analysis of data.

Accepted Manuscript

REFERENCES

1. Beebe DW. Neurobehavioral morbidity associated with disordered breathing during sleep in children: a comprehensive review. *Sleep*. 2006;29:1115-34.
2. Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, et al. Diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*. 2012;130:e714-55.
3. Chervin RD, Hedger K, Dillon JE, Pituch KJ. Pediatric sleep questionnaire (PSQ): validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. *Sleep Med*. 2000;1:21-32.
4. Downey R, 3rd, Perkin RM, MacQuarrie J. Upper airway resistance syndrome: sick, symptomatic but underrecognized. *Sleep*. 1993;16:620-3.
5. Montgomery-Downs HE, O'Brien LM, Holbrook CR, Gozal D. Snoring and sleep-disordered breathing in young children: subjective and objective correlates. *Sleep*. 2004;27:87-94.
6. Gozal D, O'Brien L, Row BW. Consequences of snoring and sleep disordered breathing in children. *Pediatr Pulmonol Suppl*. 2004;26:166-8.
7. American Academy of Pediatrics, Section on Pediatric Pulmonology, Subcommittee on Obstructive Sleep Apnea Syndrome. Clinical practice guideline: diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*. 2002;109:704-12.
8. Kheirandish-Gozal L. Practical aspects of scoring sleep in children. *Paediatr Respir Rev*. 2006;7 Suppl 1:S50-4.
9. Spruyt K, Gozal D. Development of pediatric sleep questionnaires as diagnostic or epidemiological tools: a brief review of dos and don'ts. *Sleep Med Rev*. 15:7-17.
10. Spruyt K, Gozal D. Pediatric sleep questionnaires as diagnostic or epidemiological tools: a review of currently available instruments. *Sleep Med Rev*. 15:19-32.
11. Brockmann PE, Schaefer C, Poets A, Poets CF, Urschitz MS. Diagnosis of obstructive sleep apnea in children: A systematic review. *Sleep medicine reviews*. 2013.
12. Villa T, Miralles Torres A, Beseler Soto B. [Spanish version of the Pediatric Sleep Questionnaire (PSQ). A useful instrument in investigation of sleep disturbances in childhood. Reliability analysis]. *An Pediatr (Barc)*. 2007;66:121-8.
13. Ogden CL, Kuczmarski RJ, Flegal KM, Mei Z, Guo S, Wei R, et al. Centers for Disease Control and Prevention 2000 growth charts for the United States:

improvements to the 1977 National Center for Health Statistics version. *Pediatrics*. 2002;109:45-60.

14. Chervin RD, Weatherly RA, Garetz SL, Ruzicka DL, Giordani BJ, Hodges EK, et al. Pediatric sleep questionnaire: prediction of sleep apnea and outcomes. *Arch Otolaryngol Head Neck Surg*. 2007;133:216-22.

15. Iber C, Ancoli-Israel S, Chesson A, Quan S. The AASM Manual for the Scoring of Sleep and Associated Events: Rules, Terminology, and Technical Specifications. Westchester, Ill American Academy of Sleep Medicine. 2007.

16. EEG arousals: scoring rules and examples: a preliminary report from the Sleep Disorders Atlas Task Force of the American Sleep Disorders Association. *Sleep*. 1992;15:173-84.

17. Berry RB, Budhiraja R, Gottlieb DJ, Gozal D, Iber C, Kapur VK, et al. Rules for scoring respiratory events in sleep: update of the 2007 AASM Manual for the Scoring of Sleep and Associated Events. Deliberations of the Sleep Apnea Definitions Task Force of the American Academy of Sleep Medicine. *Journal of clinical sleep medicine : JCSM : official publication of the American Academy of Sleep Medicine*. 2012;8:597-619.

18. Liberati A, Altman DG, Tetzlaff J, Mulrow C, Gotzsche PC, Ioannidis JP, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. *J Clin Epidemiol*. 2009;62:e1-34.

19. Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, et al. Diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*. 2012;130:576-84.

20. Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proc Am Thorac Soc*. 2008;5:242-52.

21. Brockmann PE, Bertrand P, Pardo T, Cerda J, Reyes B, Holmgren NL. Prevalence of habitual snoring and associated neurocognitive consequences among Chilean school aged children. *International Journal of Pediatric Otorhinolaryngology*. 2012.

22. Urschitz MS, Brockmann PE, Schlaud M, Poets CF. Population prevalence of obstructive sleep apnoea in a community of German third graders. *Eur Respir J*. 2010;in press.

23. Carroll JL, McColley SA, Marcus CL, Curtis S, Loughlin GM. Inability of clinical history to distinguish primary snoring from obstructive sleep apnea syndrome in children. *Chest*. 1995;108:610-8.

24. Li AM, Cheung A, Chan D, Wong E, Ho C, Lau J, et al. Validation of a questionnaire instrument for prediction of obstructive sleep apnea in Hong Kong Chinese children. *Pediatr Pulmonol*. 2006;41:1153-60.

25. Brouillette R, Hanson D, David R, Klemka L, Szatkowski A, Fernbach S, et al. A diagnostic approach to suspected obstructive sleep apnea in children. *J Pediatr*. 1984;105:10-4.
26. Spruyt K, Gozal D. Screening of Pediatric Sleep Disordered Breathing: A Proposed Unbiased Discriminative Set Of Questions Using Clinical Severity Scales. *CHEST*. 2012.
27. Brockmann PE, Urschitz MS, Schlaud M, Poets CF. Primary snoring in school children: prevalence and neurocognitive impairments. *Sleep & breathing = Schlaf & Atmung*. 2012;16:23-9.
28. Kennedy JD, Blunden S, Hirte C, Parsons DW, Martin AJ, Crowe E, et al. Reduced neurocognition in children who snore. *Pediatr Pulmonol*. 2004;37:330-7.
29. Lin CH, Guilleminault C. Current hypopnea scoring criteria underscore pediatric sleep disordered breathing. *Sleep Med*. 2011;12:720-9.
30. O'Brien LM, Mervis CB, Holbrook CR, Bruner JL, Klaus CJ, Rutherford J, et al. Neurobehavioral implications of habitual snoring in children. *Pediatrics*. 2004;114:44-9.
31. Calhoun SL, Vgontzas AN, Fernandez-Mendoza J, Mayes SD, Tsaoussoglou M, Basta M, et al. Prevalence and risk factors of excessive daytime sleepiness in a community sample of young children: the role of obesity, asthma, anxiety/depression, and sleep. *Sleep*. 2011;34:503-7.
32. Gozal D, Kheirandish-Gozal L. Obesity and excessive daytime sleepiness in prepubertal children with obstructive sleep apnea. *Pediatrics*. 2009;123:13-8.
33. Lopes C, Esteves AM, Bittencourt LR, Tufik S, Mello MT. Relationship between the quality of life and the severity of obstructive sleep apnea syndrome. *Braz J Med Biol Res*. 2008;41:908-13.

Fig. 1. Receiver operating characteristic (ROC) curve for PSG total scale and subscale

TABLES

Table 1. Demographic and clinical characteristics of the study sample, cases, and controls.				
	Total N=83	Primary snorers N=48	OSA N=35	Variable skewness
Males; N (%)	53 (64)	29 (60)	24 (69)	-1.69

Age [years]; mean \pm SD	9.5 \pm 3.6	9.7 \pm 3.7	9.3 \pm 3.3	-0.92
BMI [kg/m ²]; median (min – max)	18.1 (12.5-33.7)	17.8 (13.5-32)	18.9 (14.7-33.7)	1.26
Obesity; N (%)	15 (20.5)	5 (11.9)	10 (32.3)	1.01
TST [minutes]; median (min – max)	456 (212-552)	461 (212-551)	441 (259-552)	1.17
Sleep latency [minutes] ; median (min – max)	27 (0 – 160)	27 (0.5 – 109)	23 (0 – 160)	1.78
Sleep efficiency [%]; median (min – max)	87.7 (54.8-99.2)	87.8 (56.5-99.2)	85.5 (54.8-98.5)	1.41
Central apnea index; median (min – max)	0.1 (0 – 2.2)	0.05 (0 – 1.2)	0.1 (0 -2.2)	2.51
Obstructive apnea index; median (min – max)	0 (0 – 7)	0 (0 – 0.3)	0.9 (0 – 7)*	4.11
Hypopnea index; median (min – max)	0.4 (0 – 19)	0.4 (0 – 0.7)	1.5 (0.1 – 19)*	3.68
AHI; median (min – max)	0.5 (0-28)	0.4 (0-0.8)	2.5 (1 - 28)*	3.72
Arousal index; median (min – max)	11.5 (0.1-42.9)	9.4 (3 - 34.7)	11.5 (0.1- 42.9)	1.11
SpO ₂ [%]; mean \pm SD	97.2 \pm 1.1	97.7 \pm 1	96.6 \pm 1.2*	-0.43
SpO ₂ nadir; median (min – max)	91 (68-96)	91 (69-96)	91 (68-96)*	-1.82
SpO ₂ desaturation by 3% index; median (min – max)	0 (0 – 11.1)	0 (0 – 9.8)	0.1 (0.1 – 11.1)*	5.11
Snoring time in PSG [% of TST] ; median (min – max)	0.8 (0 – 50.9)	0.25 (0 – 23.3)	3.5 (0 – 50.9)*	2.56

Abbreviations: AHI, apnea hypopnea index; BMI, body mass index; OSA, obstructive sleep apnea; PSG, polysomnography; TST, total sleep time; SD, standard deviation. If not otherwise stated, all results are medians (minimum-maximum). *p-values <0.05.

Accepted Manuscript

Table 2. Comparison of PSQ items between cases and controls

PSQ Item	Primary snorers N=48	OSA N=35	P - value	Loading
Usually snores	8 (16.7)	14 (40)	0.050	0.746
Always snores	4 (8.3)	14 (40)	0.002	0.876
Snores loudly	6 (12.5)	13 (37.1)	0.031	0.840
Heavy breathing	16 (33.3)	14 (41.2)	0.709	0.442
Trouble breathing	7 (14.6)	15 (44.1)	0.003	0.743
Observed apneas	5 (10.4)	14 (40)	0.007	0.749
Mouth open during day	25 (58.1)	28 (87.5)	0.009	0.643
Dry mouth on awakening	18 (41.9)	17 (53.1)	0.488	0.163
Unrefreshed in morning	26 (56.5)	9 (25.7)	0.005	0.907
Problem with sleepiness	26 (56.5)	15 (44.1)	0.213	0.258
Sleepy per teacher	20 (44.4)	12 (37.5)	0.326	0.258
Hard to wake up	22 (45.8)	21 (61.8)	0.115	0.260
Does not listen	17 (37.8)	17 (53.1)	0.135	0.293
Difficulty organizing	20 (44.4)	21 (65.6)	0.054	0.108
Easily distracted	28 (62.2)	22 (68.8)	0.365	0.036
Fidgets	23 (51.1)	23 (71.9)	0.055	0.218
On the go	13 (29.5)	15 (46.9)	0.096	0.242
Interrupts	18 (40.9)	19 (59.4)	0.087	0.209
Nocturnal enuresis	5 (10.9)	8 (23.5)	0.113	0.057
Morning headache	8 (18.6)	6 (18.2)	0.602	0.009
Delayed growth	8 (18.6)	6 (18.2)	0.602	0.107
Obesity	7 (14.6)	9 (25.7)	0.146	0.113

Abbreviations: OSA, obstructive sleep apnea; PSQ, Pediatric Sleep Questionnaire.

Results are given as N (%). P-values (significant in bold) were obtained using χ^2 .

Accepted Manuscript

Table 3. Correlations between PSQ questionnaire score and PSG indices.

PSG index	r_s	P-value
TST	-0.063	0.570
Sleep latency	0.041	0.716
Sleep efficiency	0.015	0.891
Central apnea index	0.212	0.054
Obstructive apnea index	0.121	0.051
Hypopnea index	0.311	0.001
AHI	0.313	0.004
Arousal index	0.107	0.337
Mean SpO2	-0.118	0.073
Minimum SpO2	-0.057	0.613
Snoring time in PSG	0.195	0.089

Abbreviations: AHI, apnea hypopnea index; r_s , Spearman's rank correlation; PSQ, Pediatric Sleep Questionnaire; PSG, polysomnography; SpO2, pulse oximetry derived oxygen saturation; TST, total sleep time. Significant correlations marked in bold.

Table 4. Diagnostic test accuracy results stratified by age group***PSQ cutoff >0.33***

Age group	2- 6 years	6 -10 years	> 10 years	Total sample
Sensitivity	0.571	0.714	0.786	0.714
Specificity	0.667	0.533	0.458	0.521
Positive predictive value	0.571	0.588	0.458	0.521
Negative predictive value	0.667	0.667	0.786	0.714
Positive likelihood ratio	1.714	1.531	1.451	1.491
Negative likelihood ratio	0.643	0.536	0.468	0.549
Correctly identified N(%)	57	71	79	71

PSQ subscale cutoff >0.1

Sensitivity	1.000	0.929	0.786	0.886
Specificity	0.556	0.467	0.375	0.538
Positive predictive value	0.636	0.619	0.423	0.534
Negative predictive value	1.000	0.875	0.750	0.840
Positive likelihood ratio	2.250	1.741	1.257	1.575
Negative likelihood ratio	<0.0001	0.153	0.571	0.261
Correctly identified (%)	100	93	79	89